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DOI: 10.4103/bc.bc_67_20

Vertebral artery dissection and associated ruptured intracranial pseudoaneurysm successfully treated with coil assisted flow diversion: A case report and review of the literature

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Abstract:

Dissecting intracranial pseudoaneurysms (IPs) are associated with a high incidence of rupture and poor neurologic outcomes. Lesions in the posterior circulation are particularly malignant and pose even greater management challenges. Traditional management consists of microsurgical vessel sacrifice with or without bypass. Flow diversion (FD) in the setting of subarachnoid hemorrhage (SAH) represents a reconstructive treatment option and can be paired with coil embolization to promote more rapid thrombosis of the lesion. We report a case of a ruptured dissecting vertebral artery (VA) IP successfully acutely treated with coil-assisted FD. A 53-year-old male presented with a right V4 dissection spanning the origin of the posterior inferior cerebellar artery and associated ruptured V4 IP. The patient was treated with coil-assisted FD. Oral dual-antiplatelet therapy (DAPT) was initiated during the procedure, and intravenous tirofiban was used as a bridging agent. Immediate obliteration of the IP was achieved, with near-complete resolution of the dissection within 48 h. The patient made a complete recovery, and angiography at 6 weeks confirmed total IP obliteration, reconstruction of the VA, and a patent stent. The use of FD and DAPT in the setting of acute SAH remains controversial. We believe that coil-assisted FD in carefully selected patients offers significant advantages over traditional microsurgical and endovascular options. The risks posed by DAPT and potential for delayed thrombosis with FD can be effectively mitigated with planning and the development of protocols. We discuss the current literature in the context of our case and review the challenges associated with treating these often devastating lesions.

Keywords:

Endovascular surgery, flow diversion, pseudoaneurysm, subarachnoid hemorrhage

Introduction

Dissecting intracranial pseudoaneurysms (IPs) are relatively rare lesions that are associated with a high risk of rupture and significant morbidity and mortality.^[1] Lesions result from disruption of the vessel wall and are contained by only thin friable intima.^[1] As a result, IPs are generally not amenable to clip ligation or coil embolization, as any

manipulation usually leads to disruption of the contained thrombus and potentially catastrophic bleeding.^[1-4] Thus, traditional open and endovascular techniques have relied on vessel deconstruction with or without bypass.^[1,2] When occurring in the posterior circulation, these lesions may be particularly devastating due to their inclusion of brainstem perforators and the need for advanced skull base techniques to access the pathology.

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How to cite this article: Scullen T, Mathkour M, Werner C, Zeoli T, Amenta PS. Vertebral artery dissection and associated ruptured intracranial pseudoaneurysm successfully treated with coil assisted flow diversion: A case report and review of the literature. *Brain Circ* 2021;7:159-66.

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Submission: 29-11-2020
Revised: 01-02-2021
Accepted: 30-04-2021
Published: 27-08-2021

Flow diversion (FD) technology, initially introduced for the management of large unruptured aneurysms of the petrous to communicating segments of the internal carotid artery (ICA), has been increasingly and successfully applied to off-label indications.^[4-6] In the United States, the primary experience has been with the pipeline embolization device (PED), however, a number of additional devices have now begun to enter the market.^[4] Flow-diverting stents rely on increased metal coverage to restrict flow into the aneurysm, promote thrombosis, and provide a scaffold for endothelialization across the neck.^[7-9] In addition to providing durable long-term aneurysm occlusion, these properties also allow FDs to reconstruct the vessel anatomy, thus making them useful in addressing pathology of the vessel walls.^[7-9] There is a growing body of literature highlighting the successful treatment of dolichoectasia, fusiform aneurysms, and intracranial dissections with FD.^[10-19] However, significant debate remains in relation to their application in the setting of subarachnoid hemorrhage (SAH).^[12,20,21] FD stents require at least 6 months of dual-antiplatelet therapy (DAPT) to allow for endothelialization, which has limited their utility in the setting of acute hemorrhage.^[7-9] Potential risks of DAPT following SAH include an increased risk of re-rupture and bleeding complications in the event of additional surgical procedures (i.e. external ventricular drain [EVD] placement, shunting, and decompressive craniectomy).^[22-26] Furthermore, the acute presentation does not allow for planned pretreatment with DAPT, thereby increasing the risk of thromboembolic complications.^[27-30]

The management of acutely ruptured intracranial dissections and associated IPs remains challenging.^[31] FD is a controversial treatment option, but one that has become increasingly highlighted in the literature.^[12,13] We report successful treatment of a ruptured vertebral artery (VA) V4 segment dissection and IP with coil-assisted FD (FDAC). The technical aspects of the procedure, the management of DAPT, and the existing literature are discussed.

Case Description

A 53-year-old male with hypertension developed a severe headache and right-sided neck pain on the evening prior to presentation. He awakened the following morning with the worst headache of his life and rapidly deteriorated at an outside institution requiring intubation. Computed tomography (CT) showed diffuse SAH with a predominance of blood in the posterior fossa and hydrocephalus [Figure 1]. On examination, the patient was arousable to stimulation and followed commands symmetrically.

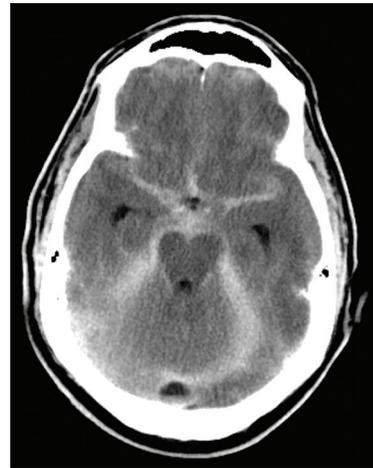


Figure 1: Noncontrast axial computed tomography of the head showing diffuse subarachnoid blood and hydrocephalus

A right frontal EVD was placed, and the patient was taken for emergent angiography. Digital subtraction angiography (DSA) demonstrated a dissection of the right VA V4 segment spanning the origin of the posterior inferior cerebellar artery (PICA) and an associated IP with a clear excrescence indicating the point of rupture [Figure 2]. Treatment proceeded with FDAC. Enteral access was gained via a nasogastric tube, and the patient was loaded with 180 mg of ticagrelor and 650 mg of aspirin. Intravenous (IV) tirofiban at 0.1 µg/kg/min was also started. A 6 Fr shuttle sheath was placed in the subclavian artery, and the right VA was selected with a Navien 072 intracranial support catheter (Medtronic, Dublin, Ireland). The IP primary sack was selected with a Duo microcatheter (Terumo, Tokyo, Japan), and a Phenom microcatheter (Medtronic, Dublin, Ireland) was advanced past the dissection into the proximal basilar artery [Figure 3]. A HydroSoft 3D 3 mm × 8 cm (Terumo, Tokyo, Japan) coil was partially deployed within the IP.

A 3.5 mm × 20 mm PED Flex (Medtronic, Dublin, Ireland) was opened across the entire dissection, jailing the coiling catheter [Figure 3]. The remaining coil was deployed and embolization was completed with additional coils, resulting in immediate obliteration of the primary IP sack [Figure 3]. Final DSA demonstrated preservation of the PICA and significant contrast stasis along the dissected segment [Figure 3]. The patient awoke neurologically intact and was maintained on daily 81 mg aspirin and 90 mg ticagrelor bid. The tirofiban drip was discontinued after 24 h, and a therapeutic P2Y₁₂ assay was confirmed. Interval DSA on postbleed day 2 confirmed a patent stent, complete obliteration of the primary sack, and minimal contrast stasis along the dissection [Figure 4].

The patient had an uncomplicated postoperative course but did require a ventriculoperitoneal shunt (VPS) on

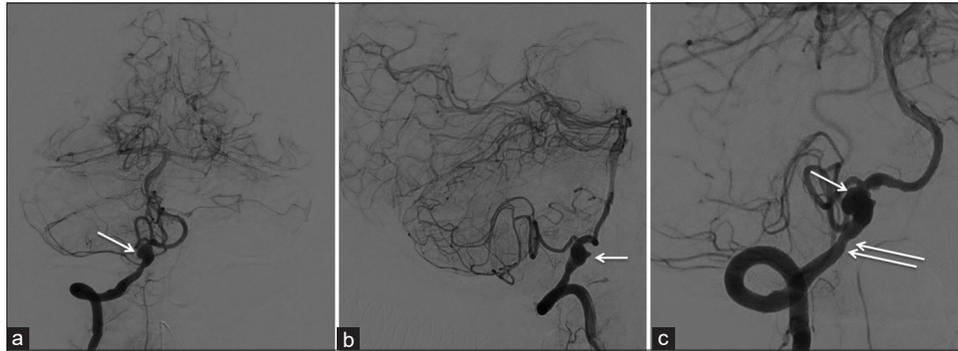


Figure 2: Right vertebral artery digital subtraction angiography anteroposterior (a) and lateral (b) views demonstrating the V4 segment dissection and associated intracranial pseudoaneurysm (arrow). (b) The excrescence (arrow), denoting the point of rupture, is seen emanating from the intracranial pseudoaneurysm. (c) Magnified lateral view again showing dissection associated narrowing of vertebral artery proximal to posterior inferior cerebellar artery (double arrows), the large ruptured intracranial pseudoaneurysm (arrow), and a second smaller sack distal to the posterior inferior cerebellar artery origin

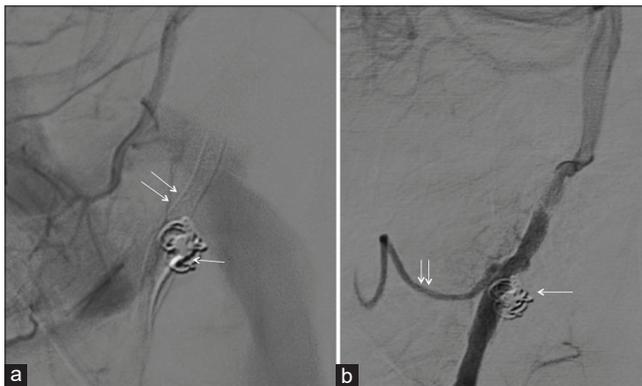


Figure 3: (a) Digital subtraction angiography lateral view demonstrating the jailed coiling microcatheter (arrow) and deployed pipeline embolization device (double arrows). (b) Immediate postcoiling run demonstrating obliteration of the intracranial pseudoaneurysm and preservation of the posterior inferior cerebellar artery and vertebral artery



Figure 4: Digital subtraction angiography anteroposterior (a and b) lateral views of the right vertebral artery 48 h postintervention. There is complete obliteration of the intracranial pseudoaneurysm and second small distal sack. (b) Minimal residual irregularity noted along the vertebral artery dissection (arrow)

postoperative day 13. Ticagrelor was discontinued 24 h prior to the shunt, while aspirin was continued. The EVD was exchanged for the proximal VPS catheter without a stylette through the existing tract to minimize the risk of catheter-related bleeding. Postoperative CT demonstrated no new hemorrhage, and ticagrelor was resumed immediately. The patient was discharged

neurologically intact, and DAPT was continued for 6 months. Four-week follow-up DSA demonstrated complete obliteration of the IP and a minimal area of vessel irregularity and contrast stasis [Figure 4], which had resolved on repeat angiography at 6 weeks [Figure 4]. Six-month DSA confirmed obliteration of the IP, resolution of the dissection, and a patent PICA [Figure 5]. Ticagrelor was discontinued, and aspirin continued indefinitely. The patient remains neurologically intact and lives independently.

Discussion

Ruptured intracranial VA dissections and IP are rare but associated with a high incidence of aneurysmal SAH (aSAH).^[6,18,19,32-35] In a series of 28 patients published by Lee *et al.*, 79% of these lesions presented with acute aSAH.^[36] There is a propensity for rebleeding, with early re-rupture rates reported between 30% and 58%.^[37] Up to 71.4% of patients suffer a rebleed within 6 h of the initial event and 93% experience a second hemorrhage within 24 h.^[38] Re-rupture is associated with extremely poor outcomes, thereby highlighting the need for emergent treatment.^[6,24,32] Traditional management has entailed deconstructive open or endovascular vessel sacrifice with or without bypass.^[1,2] In the posterior circulation, these lesions are considerably more difficult to treat due to the need for advanced skull base and bypass techniques.^[37] Ruptured VA dissections have been shown to result in overall worse outcomes, when compared to their anterior circulation counterparts.^[37] Management of perforators and the brainstem vasculature is of considerable concern, as occlusion may result in devastating neurologic deficits.^[7-9,12,20,35,39]

Reports pertaining to the use of FDAC in ruptured IPs are few,^[2,6,14,15,32] likely due to the rarity of the lesions, the associated poor outcomes, and the continued reliance on open microsurgical management.^[26] Many of these reports describe the use of FD to treat early recurrences

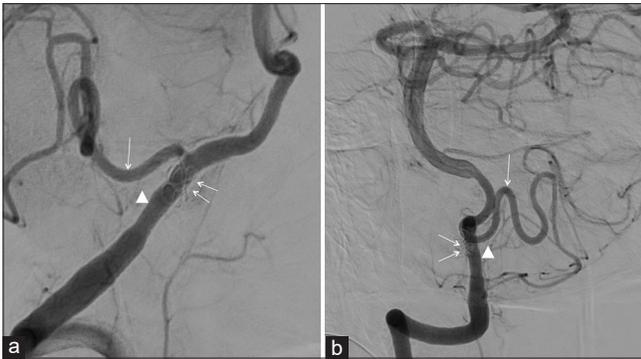


Figure 5: Six-month digital subtraction angiography lateral (a and b) oblique views of the right vertebral artery showing patent vasculature (arrow), obliteration of the intracranial pseudoaneurysm (double arrows), and resolution of the dissection (arrowhead)

after initial coil embolization, as IPs have a tendency to rapidly recur and enlarge following primary coiling.^[6,32] In these instances, FD was used as a salvage technique and performed at delayed intervals with confirmed therapeutic DAPT.^[6,32] Chiu *et al.* reported a case of an acutely ruptured dissecting V4 IP successfully treated with FDAC.^[14]

FD experience in the United States is largely based on the PED, which is approved for the treatment of unruptured large wide-necked aneurysms of the petrous to communicating segments of the ICA.^[40] The overall high rate of aneurysm occlusion, long-term durability, and excellent safety profile has driven its application to the management of challenging off-label lesions.^[7-9,12,20,35,39] The scaffolding properties that promote endothelialization and vessel reconstruction lend to its utility in treating pathology of the vessel wall.^[7-9] Due to the DAPT requirement, significant controversy continues to exist pertaining to the use of stent-assisted coil (SAC) embolization and FD technology in the setting of acute aSAH.^[34,35] In addition, posterior circulation FD has been shown to be associated with a higher risk of ischemic complications.^[18] There is, however, emerging literature supporting the safe and effective use of FD in the setting of rupture when traditional open or primary coiling techniques may have higher associated complication and lower efficacy profiles.^[1-3]

Retreatment rates among both FDAC and SAC have been shown to be significantly lower than coil embolization alone, with equivocal safety profiles in complex lesions.^[5,41,42] Likely owing to the added benefit of diversion of laminar flow secondary to increased coverage, FDAC has also been reported to have significantly lower retreatment rates as compared to SAC.^[5,41,42] Furthermore, FDAC has shown to be more cost-effective as compared to SAC, likely owing to the decreased number of coils required in dual-modality treatment. This observation is intuitive, as in SAC, the

stent exists to support and prevent prolapse of a large coil mass, which is the primary treatment modality, whereas in FDAC, the coils provide apposition and an immediate safety net for the delayed primary modality of FD. The patient in our case also presented with an aneurysm spanning the V4 segment which straddled the anterior medullary segment of the PICA, a region rich in critical myelencephalic perforators.^[43] SAC would have required a complex construct to ensure patency of the PICA and relevant perforators while simultaneously treating the entire length of diseased vessel segment.

Multiple factors must be taken into consideration when treating ruptured VA dissecting IPs, particularly the relative location of the PICA origin.^[2,6,14,18,32] Dissections proximal to the PICA may be treated with VA deconstruction if there is a sizable contralateral VA or significant collateralization through robust posterior communicating arteries.^[37] Inclusion of the PICA origin within the dissection complicates decision-making.^[37] VA sacrifice risks potential devastating infarction secondary to occlusion of PICA perforators to the brainstem and could also lead to symptomatic cerebellar infarction.^[37] These lesions can be addressed with microsurgical trapping of the diseased VA segment and revascularization of the PICA via PICA-to-PICA side-to-side anastomosis, occipital artery to PICA end-to-side anastomosis, or PICA re-implantation on a healthy VA segment.^[37] We believe, as is shown in our case, that vessel reconstruction with FD offers an additional effective treatment option for this particular subset of lesions.

Our patient suffered a right V4 segment dissection across the PICA origin with a large primary IP proximal to the vessel and second smaller irregular pouch distally. While open transcranial options were considered, FDAC was chosen for a number of reasons. First, endovascular intervention allowed for simultaneous rapid diagnosis and immediate treatment. We find this to be an important consideration in the context of extremely high rates of early re-rupture and associated poor outcomes.^[6,24,32] Second, FD reconstitutes normal angioarchitecture along the dissection.^[4] Consequently, the origin of the PICA and brainstem perforators are preserved, evidenced in our case by a lack of radiographic or clinical findings suggesting posterior circulation ischemia. Third, we found adjuvant coil embolization to immediately occlude the ruptured IP, thereby nearly eliminating the risk of early re-rupture. Repeat angiography demonstrated near-complete resolution of the pathology within 48 h and durable complete occlusion and preservation of normal vasculature at 6 months without clinical deficit.

Controversies in flow diversion in the setting of acute hemorrhage

Contemporary reports describe multiple inherent risks

when applying FD in the setting of acute rupture.^[5,6,10-14] The majority of arguments against FD in SAH center around the risks of the DAPT requirement.^[26-30] First, the patient is unable to undergo customary DAPT loading prior to the procedure and must be administered the medications during or shortly before the intervention.^[26-30] This sequence of events often precludes confirmatory platelet inhibition testing prior to FD deployment, thus risking ischemic complications.^[10,44,45] A confirmed response to DAPT is considered crucial in the prevention of acute stent thrombosis and other thromboembolic complications.^[10] Clopidogrel nonresponders, composing up to 20% of the population, are at significantly increased risk of ischemic complications when undergoing FD.^[27,28] Furthermore, clopidogrel effectiveness can be negated by proton-pump inhibitors, the use of which may be an unknown in emergent scenarios, predisposing patients to thromboembolic events.^[27,28] Second, platelet inhibition prior to complete IP occlusion risks potentially catastrophic re-re-rupture.^[27,28] Third, the 6-month DAPT requirement complicates postoperative management, as patients may require additional procedures, such as shunting, decompressive craniectomies, and tracheostomy.^[22-26]

We believe that careful planning and establishing management protocols can mitigate the risks of DAPT and allow for the safe application of FD in the setting of SAH. Perhaps most importantly, is the aggressive early management of hydrocephalus. We placed an EVD immediately upon admission and prior to intervention to avoid placing the catheter while on therapeutic DAPT. Preemptive EVD placement followed by DAPT administration has been shown to be safe and effective previously in series of SAH patients treated with SAC.^[46-49] In the event the patient requires shunting, allowing a pericatheter tract to develop over the course of 10–14 days facilitates a “soft pass” exchange of the VPS for the EVD catheter,^[24,25] thereby greatly reducing the risk of tract hemorrhage.^[50,51] This technique, along with brief discontinuation of ticagrelor for 24 h, was effectively used in our case. Temporary antiplatelet monotherapy agent at a delayed time point allows for at least early endothelialization of the stent and reduces the perioperative risk of ischemic complications.^[44]

Our patient was loaded with 650 mg aspirin and 180 mg ticagrelor via nasogastric tube in the AngioSuite following the diagnostic portion of the procedure and immediately prior to FDAC. Ticagrelor was chosen, as opposed to clopidogrel, due to the higher likelihood of a therapeutic response.^[45,52,53] To offset the risk of unpredictable absorption and response, the patient was also started on IV tirofiban to ensure immediate therapeutic platelet inhibition.^[44] Tirofiban was continued until adequate time (24 h) had passed for

therapeutic oral DAPT to take effect. Forty-eight-hour and long-term follow-up demonstrated a widely patent stent and preservation of the right VA and PICA, along with no symptoms of ischemic insult.

Multiple studies have shown stand-alone FD in the setting of rupture to be associated with a high risk of re-rupture (4%–17%) and increased morbidity and mortality.^[54] The increased incidence of re-rupture is likely secondary to two confounding factors. First, the literature demonstrates only 71% of aneurysms to immediately and completely occlude following FD alone,^[35] leaving residual aneurysm that is prone to rebleeding. Second, the administration of DAPT and/or IV tirofiban could place the patient at an increased risk of re-rupture in the immediate postoperative period.^[34,35] Ten Brinck *et al.* reported only 45% of patients undergoing stand-alone FD for ruptured aneurysms to achieve a favorable clinical outcome.^[55] Of patients treated, 11% rebled and 27% experienced a permanent neurologic deficit.^[55] A significant fraction (25%) of aneurysms treated in this study were of a dissecting etiology.^[55] McAuliffe and Wenderoth reported 2 of 11 patients suffered fatal rebleeds following FD,^[56] and a meta-analysis by Madaelil *et al.* found a re-rupture rate of 6% in patients treated with stand-alone FD.^[57]

There is a growing body of literature, particularly dedicated to ruptured large and giant aneurysms, that demonstrates coil-assisted FD to be a safe and effective treatment option.^[58-60] Chalouhi *et al.* reported only one intraoperative complication, a fatal re-rupture, in a series of 20-ruptured aneurysms treated with FD.^[60] Importantly, 30% of patients underwent adjuvant coiling. In the same meta-analysis mentioned above, Madaelil *et al.* found only 3% of aneurysms treated with coil-assisted FD to re-rupture compared to the 6% treated with only FD.^[57] In cases of dissection and IP, coil-assisted FD was found to result in significantly higher occlusion rates at extended time points without further risk of thromboembolic, hemorrhagic, or ischemic complications,^[18] and has been reported successful in ruptured IPs.^[6,14,30,32] We addressed the risk of re-rupture by coiling and immediately obliterating the primary sack and rupture point. In addition, coiling of the sack introduces thrombotic material into the false lumen and promotes more expedient thrombosis of the remaining dissection.

Differences between spontaneous and posttraumatic pseudoaneurysm

Intracranial aneurysms secondary to trauma are more commonly pseudoaneurysms than true aneurysms with intact adventitia, thus representing a common scenario in which pseudoaneurysms are encountered.^[61] Reported intracranial locations include the ICA, anterior

cerebral artery, VA, and middle meningeal artery, all of which are vulnerable to injuries as a result of being adjacent to commonly fractured skull locations.^[62] In these situations, open surgery may be more feasible in order to concurrently repair fractures or perform decompressive hemicraniectomy. However, in the case of closed head injury, pseudoaneurysms secondary to shear or rotational forces may be amenable to endovascular treatment with coil embolization or FDAC.^[62] Moreover, delayed hemorrhagic presentations can occur weeks after the initial injury, and thus, surveillance DSA should be utilized in order to visualize pseudoaneurysm growth and prevent catastrophic rupture.^[63]

Conclusion

Our patient presented with a ruptured V4 dissection that spanned the origin of the PICA and an associated IP with a clear excrescence indicating the point of rupture. While traditional microsurgical options could have been effectively employed in this case, we chose FDAC. The immediacy of treatment, the early exclusion of the ruptured IP, and the reconstruction and preservation of the diseased vessel contributed to a successful outcome. While significant controversy remains regarding the use of DAPT therapy and FD in the setting of SAH, there is an emerging body of literature that supports their application in carefully selected cases where microsurgical and traditional endovascular techniques have greater risk profiles. We believe that risk was mitigated in this case by aggressive preoperative management of hydrocephalus, coiling and obliteration of the ruptured sack, the use of IV tirofiban as a bridge to oral DAPT, and delayed shunting through a well-formed tract. Adjuvant coiling to promote timely thrombosis is key to preventing perioperative rupture. Long-term follow-up and the accumulation of experience are required to develop improved protocols for the management of these potentially neurologically devastating lesions.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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